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Laparoscopic approach to a large adrenocortical oncocytoma: A case report and review of the literature

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ABSTRACT

INTRODUCTION: Adrenocortical oncocytomas are extremely rare tumors, considered to be non-functional and of low malignant potential. Despite the great advance in laparoscopic techniques, there are extremely limited reports of laparoscopic approach of adrenocortical oncocytomas. Herein is presented a challenging case of laparoscopic approach to a large adrenocortical oncocytoma, underlining the safety and feasibility of laparoscopy in the surgical management of these extremely rare adrenal tumors.
PRESENTATION OF CASE: A 34 year-old male was referred for surgical evaluation after the incidental discovery of a large right adrenal mass, during ultrasound examination due to renal colic. Further imaging evaluation revealed a well circumscribed capsule around the mass was demonstrated, with no evidence of infiltration of the neoplasm to periadrenal tissues. The patient was scheduled for laparoscopic right adrenalectomy, running an uneventful postoperative period. Histopathology revealed the presence of an adrenal oncocytoma.
DISCUSSION: Recent studies have demonstrated that approximately one third of adrenocortical oncocytomas are associated with hormonal hypersecretion, as well as that one fifth of them demonstrate malignant biological behavior. From this point of view, there is emerging evidence in favor of the necessity of surgical excision as the treatment of choice. In spite of the progress of laparoscopic surgery, only three cases of laparoscopic excision of these tumors have been reported up to date.
CONCLUSION: Laparoscopic surgery offers a safe alternative in confronting adrenocortical neoplasms, even when the biological behavior of the tumors cannot be pre-operatively evaluated in a definite way.

1. Introduction

Oncocytic neoplasms, also known as oncocytomas, are in the majority of cases benign tumors, arising in the kidney, thyroid and salivary glands; they have also been reported in other, more rare sites including the pituitary and parathyroid glands, respiratory tract and choroid plexus.[1] Histologically, oncocytomas are characterized by the predominance of large eosinophilic cells due to the presence of densely-packed mitochondria, when examined by electron microscopy.[2] Since the first description of adrenal oncocytomas by Kakimoto et al. in 1986,[3] approximately 110 cases of these tumors have been reported in the international literature, most frequently encountered as incidental findings.[4,5]

Despite the fact that they have been traditionally considered to be non-functional and benign in their vast majority, recent data indicate that about 20% of the adrenocortical oncocytomas demonstrate elements of malignancy and, additionally, 25–30% of them appear to be associated with adrenal hormones' hypersecretion.[4] As a result, the necessity of surgical intervention has become more evident in the course of confronting these rare tumors. Surgical excision is often considered inevitable and the traditional approach to these tumors has been with open surgical extirpation. However, recent advances in laparoscopic techniques have made possible the application of minimally invasive procedures for the resection of adrenal tumors. Laparoscopic adrenalectomy offers numerous advantages over its open counterpart, including less postoperative pain, shorter hospital stays, faster return to regular activity and fewer procedure-related complications.[6,7] Laparoscopic excision of large adrenal masses has been described for malignant as well as benign neoplasms, including pheochromocytomas, metastases and aldosteronomas.[8,9] Nevertheless, up to date, there are extremely limited reports of laparoscopic resection of adrenocortical oncocytomas.[10–13] Herein, we present a very challenging case of laparoscopic approach to a large adrenocortical oncocytoma. Our case study aims to highlight the safety and feasibility of laparoscopy in the surgical management of these extremely rare adrenal tumors.

2. Presentation of case

We report the case of a 34 year-old male non-smoker, who presented with right upper quadrant and flank pain and symptoms of

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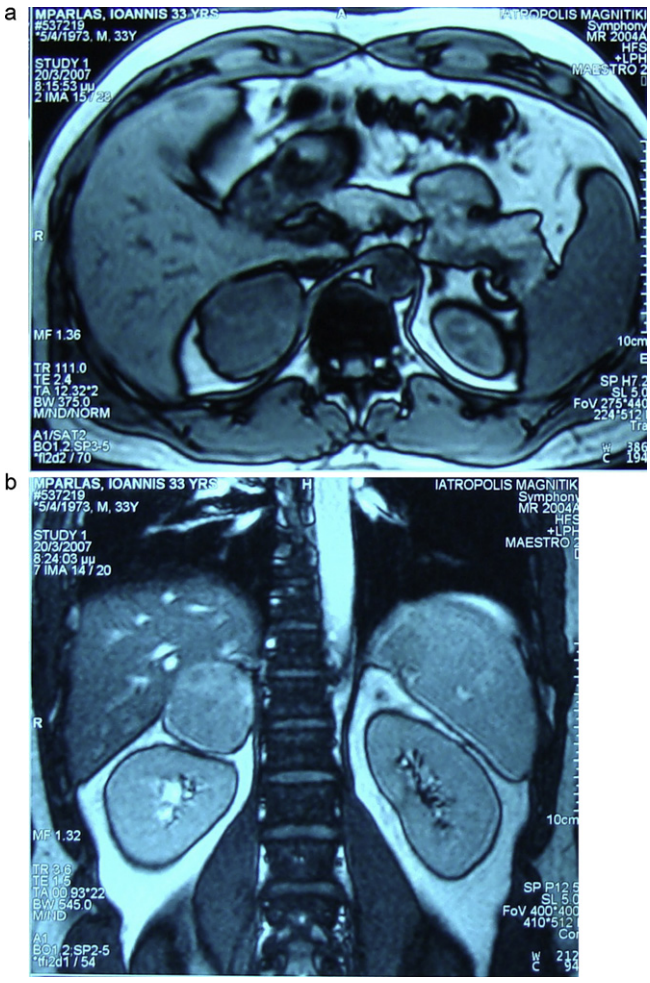

Fig. 1. Magnetic resonance imaging right adrenal tumor, measuring 6 cm. The mass has regular margins and a well circumscribed capsule, both of which are characteristics of a benign neoplasia. Note the stones in the right renal pelvis, which initially produced the symptoms of flank pain that led the patient to seek medical assistance. (A) Axial view and (B) sagittal view.

right renal colic. Renal ultrasound was performed, confirming the clinical suspicion of nephrolithiasis, with stones at the right renal pelvis, but also revealed the presence of a large right adrenal mass with mixed echogenicity. Serum electrolytes, blood urea nitrogen, creatinine and complete blood count were within normal range, while a chest roentgenogram did not reveal any obvious pathology. Urinalysis revealed two to five red blood cells per high-power field. The patient was submitted to computed tomography scan of the abdomen, which demonstrated a large suprarenal retroperitoneal tumor, 6 cm at its largest diameter, with no lymphadenopathy or any other intra-abdominal neoplastic sites. Further diagnostic workup did not reveal hypertension, headaches, palpitations or inappropriate perspiration, whereas adrenocortical hormones levels were normal. After administration of intravenous crystalloids and acetaminophen, the patient reported spontaneous passage of two stones, while the symptoms of the renal colic gradually regressed. Because of high clinical suspicion of a malignant adrenal tumor, owning to its size, an MRI scan was performed to clarify the relation of the mass with the adjacent tissues, during which the presence of a well circumscribed capsule around the mass was demonstrated, with no evidence of infiltration of the neoplasm to periadrenal tissues (Fig. 1A and B). Furthermore, tumor homogeneity and regular margins were seen, both of which added to the diagnosis of a benign neoplasm. The patient was submitted

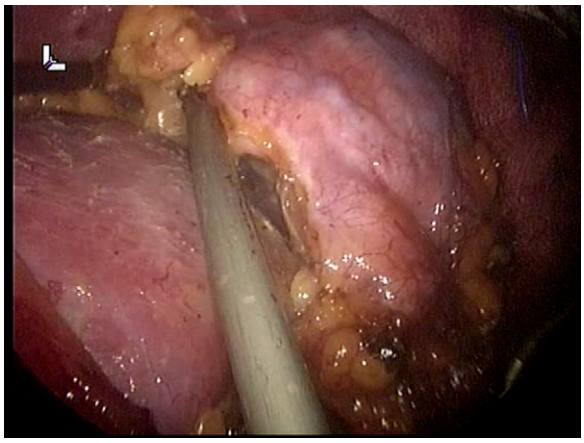

Fig. 2. Intraoperative laparoscopic view of the tumor (white thick arrow), of the perirenal fat (white thin arrow) and of the superior aspect of the right kidney (black arrow).

to laparoscopic mass excision, using the lateral transperitoneal approach. The mass was completely encapsulated and easily dissected with its perirenal fat from the liver and superior pole of the right kidney (Figs. 2 and 3). No major bleeding or hemodynamic instability events were encountered perioperatively. After an uneventful postoperative recovery period of 6 days the patient was discharged in good medical condition. During a follow-up period of twelve months after the excision of the mass, any evidence of recurrence is absent.

Gross inspection of the specimen during pathologic examination, revealed a grayish and soft mass, measuring $7.5 \times 6.5 \times 5.2$ in diameter and weighting 168 g. Cut surface revealed a well circumscribed mass with focal invasion of the capsule, no hemorrhage and necrosis and without cyst formation. Multiple tissue sections revealed a moderately thick, uniform fibrous capsule. Microscopic examination showed abundant large polygonal cells with eosinophilic cytoplasm and minimal mitotic figures. Immunocytochemical assays included positive staining for vimentin, synaptophysin and calretinin. Ultrastructurally, the neoplastic cell morphology was consistent with adrenocortical oncocytoma with the presence of myelolipoma foci. The cytoplasm of the neoplastic cells contained numerous packed round mitochondria, while other cellular organelles were scarce (Fig. 4).

Fig. 3. Gross specimen immediately after its resection.

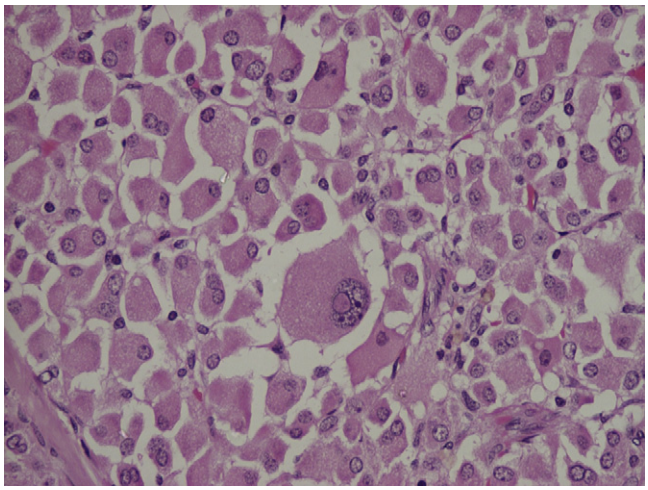

Fig. 4. Haematoxylin–eosin staining of the specimen, characteristic of the presence of abundant eosinophilic cytoplasm of the adrenal cortical cells.

3. Discussion

Increasing numbers of adrenocortical tumors are encountered in patients with no apparent clinical or hormonal adrenocortical abnormalities. These neoplasias are incidentally found with the use of contemporary imaging modalities in patients evaluated for unrelated reasons.[14] It seems justified to assume that the widespread use and availability of CT and MRI imaging techniques has resulted in this increased number of clinically "silent" adrenal tumors, like adrenocortical oncocytomas. These rare tumors are histologically characterized by cells with eosinophilic granular cytoplasm and ultrastructurally by the presence of numerous, closely-packed mitochondria.[15]

In general, these tumors have been considered to be non-functional tumors; however, 31.5% of the adrenocortical oncocytomas described up to date were associated with hormonal activity, with virilization, Cushing's syndrome and feminization being the most common endocrine clinical manifestations.[4] Moreover, with respect to the doubted malignant potential of these tumors, it is evident that adrenocortical oncocytomas demonstrate malignant behavior in approximately 22% of patients.[4] These findings reinforce the concept that adrenocortical oncocytomas, although not definitely diagnosed preoperatively, must be surgically resected. The system of Weiss, based on nine histological findings, has been widely used for classifying adrenal tumors as benign or malignant.[16] The above mentioned oncocytic tumor had abundant eosinophilic cytoplasm, as well as vascular and capsular invasion, all of which represent histologic criteria of the Weiss classification system. According to this system, the resected tumor should be considered malignant. Nevertheless, in the twelve-month postoperative follow-up, there was no evidence of local recurrence or distal metastasis.

The surgical management of adrenal tumors has been traditionally involving open surgical approach. However, recent advances in endoscopic techniques have allowed the performance of an increasing number of laparoscopic adrenalectomies. Retrospective comparison studies have consistently shown the laparoscopic approach to have less morbidity and patient discomfort, quicker patient recovery and shorter hospitalization duration, compared to open adrenalectomy.[8,17] Nevertheless, laparoscopic resection of adrenal tumors measuring more than 6 cm and/or potentially malignant adrenal tumors still remains a matter of scientific debate.[18] The major concern is that laparoscopic resection of large adrenal tumors may result in inadequate removal or capsular disruption of a malignant tumor and an increase in the risk of a local or port-sit recurrence of the disease. We suggest that laparoscopic approach to adrenal tumors can be safely performed, especially when the preoperative CT and MRI findings reveal a well-encapsulated tumor, with no evidence of invasion into surrounding tissue and no regional adenopathy, as in the above mentioned case.

4. Conclusion

Although there have been various reports for laparoscopic excision of non-functional adrenal masses, our literature review revealed only three previous reports of laparoscopic confrontation of adrenocortical oncocytomas.[10,11,13] The intact capsule of the removed tumor, the absence of perioperative adverse events, the rapid recovery of the patient and his return to regular activities, as well as the fact that no evidence of recurrence has come up after 12 months of follow up, concur to the efficacy and safety of the procedure.

Conflict of interest statement

None.

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None.

Ethical approval

The authors confirm that written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contributions

PK, EL, CS and GG were involved in drafting the manuscript. NK, DK, IM and AM were involved in collecting imaging and histopathological material, reviewing the literature and critically revising the manuscript. All authors read and approved the final paper.

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